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CYSTS IN A BRUNNER'S GLAND HAMARTOMA: A CLUE TO DIAGNOSIS

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The appearance of cystic Brunner's gland hamartomas (BGHs) on computed tomography (CT) or magnetic resonance imaging (MRI) has only been reported in a very small number of cases. Imaging diagnosis of cystic BGHs is usually difficult. We present a case of cystic BGH and characterize it in conjunction with previously reported cases. We found that the cysts of BGHs are smaller than those of other cystic duodenal lesions. The presence of cysts in BGHs can limit the differential diagnosis to cystic duodenal lesions, and our observations may assist others in the discrimination of cystic BGHs from other cystic duodenal lesions.

Key-words: Brunner's gland – Hamartoma – Duodenum — Cyst.

Brunner's gland hamartomas (BGHs) are uncommon duodenal lesions, and account for approximately 5% of all duodenal masses (1). BGHs contain cysts on rare occasions, and only a very small number of cases of cystic BGH with accompanying computed tomography (CT) or magnetic resonance imaging (MRI) documentation have been reported (2-4). Imaging diagnosis of BGHs is usually a challenge, due to their nonspecific appearance. The presence of cysts can limit the differential diagnosis to cystic duodenal lesions. Characterization of the cysts in BGHs may facilitate their differentiation from other cystic duodenal lesions. Herein, we present a case of duodenal cystic BGH and review the cases reported to date, to determine the differences between the cysts of BGHs and those of other cystic duodenal lesions.

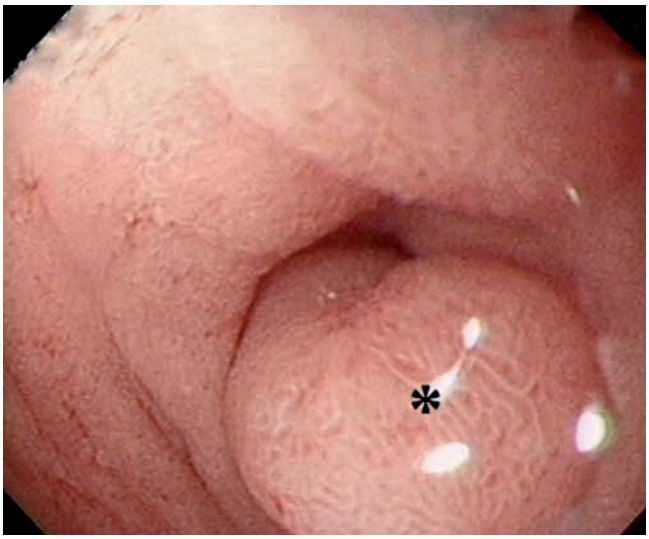

Fig. 1. — Endoscopic findings. A submucosal tumor with intact mucosa (asterisk) in the second portion of the duodenum causes luminal narrowing.

Case report

A 39-year-old man who had been experiencing postprandial epigastric pain and vomiting for three days visited our emergency department. The results of his physical examination, laboratory tests, and abdominal sonography were unremarkable. An endoscopic examination of the upper gastrointestinal tract revealed a shallow gastric ulcer and a submucosal tumor in the second portion of the duodenum causing marked luminal narrowing (Fig. 1). Contrast-enhanced abdominal CT showed a 2.2 × 4.5 cm mass with multifocal, cyst-like low densities occupying the second portion of the duodenum (Fig. 2). An endoscopic biopsy of the duodenal tumor was performed and pathologic examination revealed that the cells and lobules of the Brunner's glands were enlarged, but otherwise normal in appearance with no cellular atypia (Fig. 3). On the basis of the histologic findings, a diagnosis of a Brunner's gland hamartoma was made. During hospitalization, oral intake was withheld and esomeprazole and metoclopramide were administered intravenously. Three days after admission, his abdominal pain and vomiting resolved, and the patient was discharged from the hospital. Surgical excision of the duodenal hamartoma was not performed. The patient has followed up twice in the outpatient department in the 1 month since his discharge, and remains symptom-free.

Discussion

Brunner's glands are normally located in the duodenal submucosa, and their concentration gradually decreases towards the distal part of the duodenum. These glands produce an alkaline secretion to protect the duodenum from injury by gastric acid and optimize the pH for pancreatic enzymes. The pathogenesis of BGHs is unknown. BGHs are glandular hyperplasias or hamartomas, rather than true neoplasms. Because these lesions do not have features of cellular atypia, the terms Brunner's gland "hyperplasia" or "hamartoma" are preferable to "adenoma." The distinction between Brunner's gland hyperplasia and Brunner's gland hamartoma is arbitrary. According to the guidelines used at the Armed Forces Institute of Pathology (Washington DC, USA), if the lesion is less than 5 mm in size it is called a hyperplasia, and if it is greater than 5 mm it is called a hamartoma (5).

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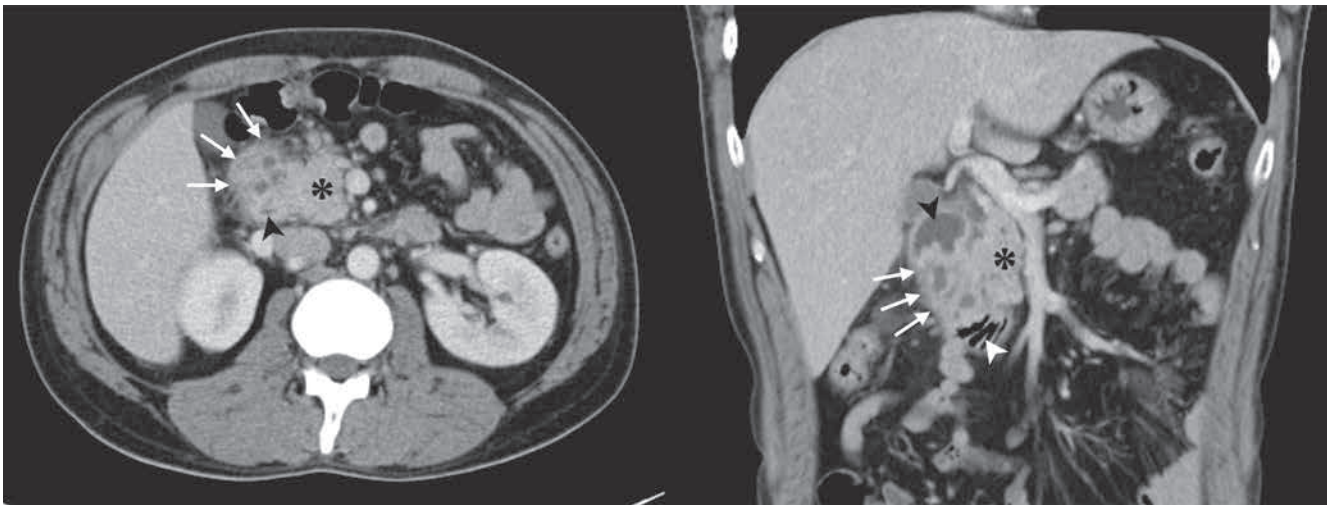

Fig. 2. — Axial and coronal contrast-enhanced CT images. A mass (white arrows) in the second portion of duodenum contains multiple small cysts.
Duodenal lumen: black and white arrowheads, pancreas: asterisks.

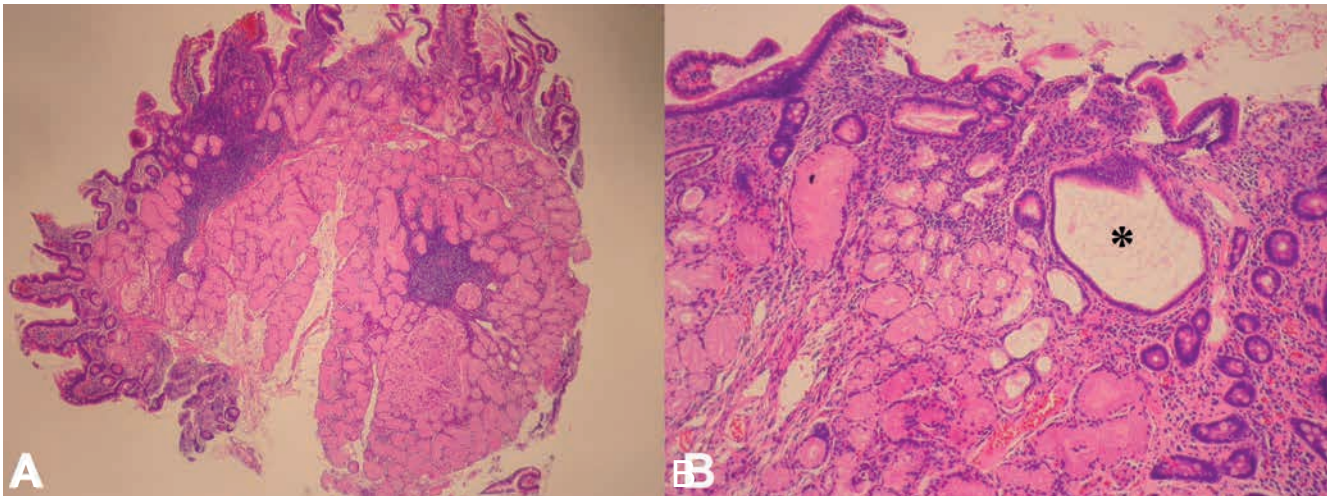

Fig. 3. — Photomicrographs of the BGH (hematoxylin and eosin, original magnification ×80 in A and ×200 in B) shows proliferation of Brunner's glands and cystic dilatation of a gland (asterisk in B).

BGHs may be symptomatic, and symptomatic lesions are often larger. The most common presentations in symptomatic patients are gastrointestinal hemorrhage (37%) and obstructive symptoms (37%) (6). In a series of 27 patients, 70% of BGHs were located in the duodenal bulb, 26% in the second portion of the duodenum, and 4% in the third portion, and most (89%) were pedunculated (6).

Cysts can only be identified in BGHs via CT or MRI on rare occasions. To our knowledge, only 4 cases demonstrating the CT or MRI appearance of cystic BGHs have been reported in the English-language literature to date (2-4). These cysts are dilated acini or ducts of Brunner's glands. The CT findings of the previously reported cases and the current case are summarized in Table I. The smaller hamartomas (< 2 cm) contain a solitary cyst, and the larger ones (≥ 4 cm) manifest as a soft tissue mass containing multiple cysts. These cysts are round, ovoid, or elongated in shape. We found that the short axes of all cysts in these BGHs do not exceed the diameter of the abdominal aorta at the same level, in axial images. Diagnosis of BGHs is usually difficult, due to nonspecific imaging findings. However, the presence of cysts is helpful in diagnosis because the differential diagnosis is limited to cystic duodenal lesions. Cysts in BGHs differ from those in other cystic duodenal lesions. Duplication cysts and lymphangiomas are thin-walled cystic masses without mural nodules or soft tissue components. These cysts are larger than those in cystic BGHs (7, 8). In contrast with conventional soft tissue and central nervous system schwannomas, cysts are unusual in gastrointestinal schwannomas (9). If present, they are fewer in number and larger than those in BGHs of similar size (4, 10). Cystic change is not uncommon in gastrointestinal stromal tumors (GISTs),

Table I. — Summary of the CT findings of cystic BGHs in the current case and the literature.

Case	Age/Sex	Size of tumors (cm)[a]	Number of cysts	Reference
1	39/M	4.5	7	Current case
2	30/M	4.0	≥ 10[c]	(2)
3	49/F	1.8	1	(3)
4	66/F	6.0	≥ 5[c]	(3)
5	48/M	< 2.0[b]	1	(4)

[a] Largest dimension.
[b] The exact size of the BGH is not available in case 5. However, the largest dimension of the hamartoma approximates the diameter of abdominal aorta, and the diameter of abdominal aorta is estimated to be less than 2 cm.
[c] Because limited images are available in these cases, exact number of cysts cannot be determined.

and is more commonly seen in large tumors. Cysts in GISTs are significantly larger than those in BGHs (11, 12). The duodenum is an uncommon site for pancreatic pseudocyst formation. In the cases reported by McCowin and Federle (13), these pseudocysts were solitary and larger than the cysts of BGHs, and in all patients, CT revealed other evidence of pancreatitis in addition to duodenal pseudocysts. Cystic dystrophy in heterotopic pancreas (CDHP), most commonly located in the second portion of the duodenum, has a similar appearance to cystic BGHs. These cysts are multiple and small, and are very similar to those seen in larger cystic BGHs. However, inflammatory changes such as periduodenal edema, periduodenal effusion, and enlarged lymph nodes are observed in most cases with CDHP (14).

Conclusion

The size of cysts is the key to diagnosing cystic BGHs. The cysts in BGHs are smaller than the cysts of other cystic duodenal lesions, except CDHP. When a mass containing a small solitary cyst or multiple small cysts is found in the first and second portion of the duodenum, without evidence of periduodenal inflammatory change or pancreatitis, cystic BGH should be considered.

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